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#### **Case Report**

# Spontaneous Adrenal Haemorrhage in Early Twin Pregnancy: A Case Report of Successful Conservative Management

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### Abbreviation

SAH: Spontaneous Adrenal Haemorrhage; MRI: Magnetic Resonance Imaging.

#### Introduction

Spontaneous adrenal haemorrhage (SAH) in pregnancy is a rare condition. Given the absence of a specific clinical presentation, SAH should be considered in the differential diagnosis of abdominal or flank pain and signs of shock at different gestational age. Any delay of the diagnosis is potentially life threatening for both mother and fetus by adrenal crisis if bilateral or in case of massive bleeding.

In this article, we present a rare case of unilateral SAH in early pregnancy with successful conservative management.

#### **Case Report**

A 31-year-old nulliparous woman, in the 15 weeks of gestation, was referred to our emergency department with intense left flank pain, radiating to the left upper quadrant abdominal, worsening with deep respiration, accompanied by vomiting. The pain started 4 weeks ago and was progressive recently.

On admission, the patient's vital signs showed blood pressure of 100/60 mmHg, pulse rate of 128 beats per minute, tachypneic of 21 breaths per minute with oxygen saturation of 100 % on air room. The patient was afebrile (37,1 C°). The fetal heart rate was normal for both of the twins.

The previous medical history was unremarkable and there was no history of major surgery, recent abdominal trauma or underlying illness. The patient denied any use of drugs or anticoagulants.

At initial physical examination, there was tenderness in left upper quadrant. The uterus found soft, the cervix closed and no vaginal bleeding was present.

The complete blood count showed the following results:

#### Abstract

Spontaneous adrenal haemorrhage in pregnancy is an extremely rare condition. Herein we present an additional case of unilateral spontaneous adrenal haemorrhage in a 31-year-old pregnant woman at the 15 weeks of gestation with successful conservative management and a safe vaginal delivery.

Keywords: Adrenal Haemorrhage; Pregnancy; Spontaneous

hemoglobin 6,8g/dl, erythrocyte 3,4 106/uL, hematocrit 29%, leucocytes 7,6 103/uL, thrombocytes 312 103/uL. Coagulation test, liver function test, renal function test, C-reactive protein and lipase were all within normal limits.

Prior to imaging, patient's general condition established at intensive care unit with blood transfusion (3 units) and 1000 ml of colloid intravenous infusion. Intravenous narcotic were prescribed to control the pain.

The patient's vital signs improved, her blood pressure increased to 110/80 mmHg, pulse rate decreased to 86 beats per minute and hemoglobin level drooped to 9.7g/dL. The fetal heart rate was still normal for both of the twin.

Transvaginal ultrasound showed a live intrauterine pregnancy with a normal appearing anterior placenta and appropriate fetal growth. Abdominal ultrasound showed a well defined and lobulated heterogeneous mass of  $14 \times 13$  cm behind the left kidney. The left adrenal haemorrhage was suspected.

Considering the stable condition of the patient, she was admitted for abservation. One day later, an abdominal ultrasound was repeated and showed no change in size of the mass.

The adrenal function was tested in blood (total cortisol) and in 24hour urine collection (free cortisol, metanephrine, normetanephrine) and were normal.

Because of gestation, an abdominal magnetic resonance imaging (MRI) was scheduled and showed a well-limited round mass of 12,4×13,3×15,6 cm in the left adrenal region with no recognize able left adrenal gland. The mass was heterogeneous with a hyperintense signal on the T1-weigted and T2-weigted images. Also, the mass was lobulated with a low signal on the T1-weighted images and high signal on the T2-weighted images in some areas, which consist with recurrent bleeding or hematoma (Figure 1). The diagnosis was a spontaneous adrenal haemorrhage in different stages.

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Figure 1: MRI results of left adrenal hemorrhage.

The patient was monitored with serial hemoglobin assessment and remained clinically stable. After 12 days, she was discharged with no change in size of the adrenal hematoma on abdominal ultrasound.

The abdominal ultrasound was carried out every month which showed a progressive decrease in size of the hematoma with no other abnormalities. The main concern was the risk of re-bleeding, and there is no consensus regarding the mode of delivery.

Based on patient's preference, she returned at 37 weeks of gestation, for induction of labor. She had an uncomplicated vaginal delivery of two females neonates in good health. She recovered well and she was discharged 3 days later with an interval imaging study to assess the complete resolution of the adrenal hematoma. Also, a serial hemoglobin measurement as well as assessment of adrenal function were planned.

## **Discussion**

Autopsy figures for adrenal haemorrhage from all causes range from 0.14% to 1.1%. [1,2] The incidence in pregnancy is unknown [1-3].

The causes of adrenal haemorrhage include mainly sepsis, trauma, anticoagulation or coagulopathy such as antiphospholipid syndrome, major surgery, adrenal adenoma or neoplasma, severe stress and pregnancy [1].

Only a few cases of adrenal haemorrhage in pregnancy have been reported at different gestational ages. Pregnancy related condition that could increase the risk of mostly bilateral adrenal rupture is preeclampsia, spontaneous abortion, antepartum or postpartum haemorrhage [1]. Uncomplicated pregnancy is rarely associated with unilateral adrenal haemorrhage [1,3].

A mechanism of SAH is mostly thought to be adrenal vascular rupture and central venous thrombosis. [3,4] The high-risk factors of increased arterial blood supply to adrenal gland with limited veinous drainage, physiological adrenal cortex hyperplasia and hypercoagulative status during pregnancy may cause haemorrhage [1,2].

SAH can be asymptomatic or symptoms can be non-specific in pregnancy, and should be considered in the differential diagnosis of abdominal or flank pain at different gestational age, because, if unrecognized, it may lead rapidly to death for both mother and fetus by adrenal insufficiency or in case of massive blood loss [1,2,6].

The standard laboratory evaluation is not helpful in establishing the diagnosis [1].

The abdominal ultrasound, typically the first imaging study in pregnancy, can suggest an adrenal haemorrhage (heterogeneous mass in the adrenal region), but the MRI is the most accurate imaging modality for the diagnosis and is needed to exclude an underlying lesions such as pheochromocytoma as the cause of the haemorrhage which may require surgical resection [1,5,6].

Surgical, interventional and conservative treatments are the main management of SAH in pregnancy, which depends upon the age of gestation, amount of bleeding, imaging findings and adrenal function.

Surgical intervention may be warranted in an unstable patient, in massive haemorrhage or when an underlying carcinoma is suspected [1,5]. Radiological intervention can also be a viable alternative in patients who are unstable or medically unfit for major surgery. If the haemorrhage is less severe and the patient is hemodynamically stable, the conservative management can be appropriate during pregnancy [1,6].

Preterm delivery may be indicated if a patient is unstable, worsening or if adrenal haemorrhage is associated with severe preeclampsia or eclampsia [6].

There is scant literature available on optimal mode of delivery, but in a stable patient, vaginal delivery can be safely undertaken as demonstrated in the reported case [6].

Surveillance in the postpartum period is required in all cases, because the stress of delivery could cause recurent adrenal haemorrhage.

#### Conclusion

During pregnancy, SAH is a very rare condition and potentially fatal. The management is multidisciplinary. An expectant management may be adequate in stable patients, with minimal risk for patient and fetus.

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